Prepubertal labial adhesions: Evaluation of a referral population

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OBJECTIVE: The purpose of this study was to assess patient demographics, clinical presentation, response to previous treatment, and current evaluation of prepubertal girls referred for gynecologic care of labial agglutination.

STUDY DESIGN: A retrospective chart review of all prepubertal female patients with labial agglutination referred from 1996 through 2001.

RESULTS: Twenty-three girls were diagnosed with labial agglutination during the review period. All of the diagnoses were made by a primary care provider. At the time of referral, most patients had received medical therapy, but had not obtained resolution of their labial agglutination with topical estrogen. Twenty-two of the 23 patients required manual separation of the labial adhesions. Findings most frequently revealed a pinpoint opening and thick (3-4 mm) adhesions with >90% of the vestibule adhesed in 21 of 23 girls. Nine of 23 girls had recurrence of adhesions. Four girls required a repeat manual separation because of recurrent thick adhesions. One of 4 girls required a third manual separation. Five of the 9 recurrences were treated successfully with topical estrogen.

CONCLUSION: Gynecologists who treat patients with labial agglutination frequently may encounter children for whom medical treatment has failed. Patients whose condition does not respond to topical therapy may have thick adhesions and require manual separation. (Am J Obstet Gynecol 2002;187:327-32.)

Key words: Labial agglutination, labial adhesions

Labial adhesions in prepubertal girls may be an asymptomatic incidental finding or may be detected while symptoms (including urinary retention, urinary tract infection, altered urinary stream, or pain with activity) are being investigated.

The incidence of labial agglutination is most frequent in younger prepubertal girls (aged 3 months to 6 years) with a peak incidence at 13 to 23 months of age.[1,2] Although adhesions occur less commonly after age 6, adhesions beginning at any age may persist or recur until puberty. They have been documented to occur in 0.6% to 3.0% of girls but may actually occur more frequently, since asymptomatic adhesions may go undetected.[2] Most of the adhesions resolve or do not occur after endogenous estrogen production at puberty. Those adhesions that occur after puberty are customarily the result of surgical procedures (such as episiotomy) or may be associated with vulvar trauma.

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The cause of labial adhesions is uncertain, although conditions that inflame or irritate the vulva (eg, vulvovaginitis, recurrent diarrhea, and dermatologic disorders) are suspected.[2]

The diagnosis of labial agglutination is made by visual inspection of the genitalia.[1] The visualization of a midline "raphe" excludes other diagnoses, such as genital anomalies or an imperforate hymen.

The treatment of asymptomatic labial agglutination is controversial. In these patients, observation with time may lead eventually to spontaneous resolution, especially when estrogen production begins with the onset of puberty. The presence of symptoms that indicate urinary tract infection, urinary retention or obstruction, or altered behavioral patterns and pain with activity, however, dictates the need for intervention. Virtually complete agglutination should also be considered for treatment, because the development of urinary symptoms in this clinical setting would preclude the ability to perform a urethral catheterization.[3]

Social forces such as family concern about genital appearance or caretaker concerns about normal anatomic findings versus conditions of abuse may also stimulate the parents' desire for treatment.

First-line treatment with estrogen cream is recommended; regimens vary in detail but involve several weeks of application, usually twice daily. Generally, the cream is applied with gentle traction to the site by the provider or the patient herself, or a cotton-tipped swab is used to apply the medication against the midline raphe.

Table I. Patient demographics: labial adhesions

Patient	*Age at referral*	*Race*	*Socioeconomic status*	*Age at initial diagnosis*
1	23 mo	African American	Medicaid	3 mo
2	21 mo	White	Medicaid	5 mo
3	20 mo	White	Private insurance	Unknown
4	18 mo	White	Private insurance	Unknown
5	19 mo	African American	Medicaid	4 mo
6	3 y	White	Private insurance	24 mo
7	4 y	White	Private insurance	Unknown
8	2 y	White	Medicaid	Unknown
9	3 y 11 mo	African American	Medicaid	Unknown
10	16 mo	African American	Medicaid	6 mo
11	3 y 10 mo	African American	Medicaid	Unknown
12	3 y 1 mo	African American	Medicaid	Unknown
13	10 mo	African American	Medicaid	3 mo
14	15 mo	White	Private insurance	Unknown
15	2 y 9 mo	White	Private insurance	Unknown
16	4 y 1 mo	Hispanic	Private insurance	9 mo
17	16 mo	White	Medicaid	4 wk
18	3 y	White	Private insurance	Unknown
19	4 y	White	Private insurance	12 mo
20	6 mo	White	Private insurance	Unknown
21	6 mo	Hispanic	Private insurance	Unknown
22	9 mo	White	Private insurance	6 mo
23	3 y 6 mo	White	Private insurance	Unknown

Response to medical therapy may be predicted by a short duration of adhesions, the elimination of irritating forces (moving from diapers to toilet training), or the appearance of a translucency in the medium raphe, which indicates the presence of very thin adhesions. A failure to respond to medical therapy with gentle traction requires consideration of other options, which include in-office treatment with manual separation after topical anesthesia or separation in an outpatient setting or surgical suite with sedation.[4,5] Topical preparations that may be helpful include lidocaine 2% (Xylocaine Jelly), lidocaine 5% (Xylocaine Ointment), and lidocaine 2.5% and prilocaine 2.5% (EMLA Cream).

Manual separation with hands or a cotton-tipped swab may be attempted after 15 to 30 minutes of topical anesthesia.[4] Thick adhesions may not be resolved without some discomfort because of the minimal depth of penetration of these products. Repeated applications may be required.

Absence of a translucent raphe or long-standing labial adhesions may be predictive of thicker adhesions. These may not respond to topical estrogen and may require manual separation. When separated, these may reveal adhesions 3 to 4 mm.[6]

Manual separation in a forceful manner without anesthesia is contraindicated because of the pain and emotional trauma to the patient and the development of raw edges that may lead to recurrence of the adhesions. Future patient cooperation is also compromised. Flavored midazolam elixir is an alternate outpatient sedative that may be combined with topical anesthesia.[5] Separation of labial adhesions in an outpatient surgical suite is usually accomplished with sedation or general anesthesia.

Care after the separation is important. Treatment is accomplished initially with topical estrogen cream for 1 to 2 weeks, which is followed by 6 to 12 months of daily application of a bland emollient (white petroleum jelly).

Material and methods

A retrospective chart review of all prepubertal girls referred for care of labial agglutination from 1996 through 2001 was conducted. Information was obtained by a review of available referral records, current charts, patient and parent interviews, and office examinations. Demographic information collected included the age at time of referral visit, race, method of payment (private insurance or Medicaid), and determination of the onset of symptoms (age at diagnosis) (Table I).

All patients had received the diagnosis of labial agglutination from their primary care provider (a pediatrician or family practitioner) and had undergone treatment with a topical estrogen preparation. Treatment regimens varied widely in duration because of practitioner individuality and patient tolerance.

The charts that were reviewed were coded by ICD-9 code 752.49. The charts were excluded if patients were not prepubertal.

After office examination, adhesions were described as filmy and translucent, or thick and without a transparent midline raphe. Also, adhesions were viewed as involving some or all of the vestibule. When all of the vestibule was involved, only a tiny or pinpoint opening allowed urine outflow.

Symptoms	*Side effects of topical estrogen*
Urinary tract infection, recurrent vulvitis	
Dysuria	
Dysuria	Pain and erythema with estrogen cream
Dysuria	Pain and erythema with estrogen cream
None, medical treatment requested by family	Pain and erythema with estrogen cream
Discomfort with activity	
Discomfort with activity	
Family requested medical treatment	
Discomfort with activity, dysuria	Pain and erythema with estrogen cream
Intermittent urinary retention, foul vaginal discharge	
Urinary tract infection	
Discomfort with activity	
Family requested medical treatment	
Family requested medical treatment	
Altered urinary stream, unable to toilet train	Breast "budding" with estrogen cream
Discomfort with activity	Breast "budding" and increased body hair with estrogen cream
Family requested medical treatment	
Family requested medical treatment	
Discomfort with activity	
Urinary retention	
Family requested medical treatment	
Family requested medical treatment	
Altered urinary stream	

Results

Eight of 23 patients were referred because of abnormal genital findings. These patients were referred for further consideration and treatment at the family's request after medical therapy had failed. Many of these children, whose family requested further treatment, were in daycare settings outside the home where the genital appearance had been repeatedly brought to the family's attention and the child's medical findings regarding past potential sexual abuse or genital anomalies had been of concern to the caretakers.

Seven of 23 patients were referred for further care because of "discomfort." Their history included verbal complaints of pain with activity, or avoidance of activities, particularly those with straddle positions (tricycle, monkey bars, see-saw). Other symptoms included pulling at panties or diapers.

Three patients had dysuria (pain with urination). Two patients had urinary retention, two patients had a urinary tract infection at the time of diagnosis, and two patients had a history of an altered urinary stream that interfered with toilet training. In patients with an altered urinary stream, the urine exited the vestibule at an angle wetting the child's panties or exiting over the toilet seat at the time of voiding. Several families also noted dribbling or dripping of urine after completion of voiding (presumably from urine that was retained initially in the vagina).

Two patients were reported to have a recurrent vulvitis or foul vaginal discharge. It could not be determined whether these symptoms preceded or followed the development of the labial adhesions because, in these two patients, evaluation of the symptoms led to the initial identification of the labial agglutination.

One set of twins developed labial agglutination at similar ages. One twin's symptoms and severe dysuria led to the evaluation of the other twin. Another patient had a younger sister with labial adhesions, but an evaluation of the younger sister, who was age 3.5 months at the time, revealed only a small agglutination in the posterior vestibular. This younger sister was asymptomatic and was not included in the study because she was seen incidentally at the time of her older sister's visits.

Twenty of 23 patients, at the time of referral, had >90% agglutination of the vestibular opening. Most had only a pinpoint or 1- to 2-mm opening in the agglutinated area through which the patient's urine exited. The remaining 3 patients had a slightly larger opening, although no more than >20% of the possible area. Openings were located over the urethral orifice. None of the patients exhibited a translucency in the midline raphe.

Twenty-two of 23 patients were taken to the outpatient surgical suite for separation with general anesthesia. The Department of Obstetrics and Gynecology at the University of South Carolina School of Medicine/Palmetto Richland Memorial Hospital has developed a working relationship with the Department of Anesthesia regarding the treatment of these patients. These customarily healthy patients are treated with mask anesthesia alone without preoperative blood work or intravenous placement. This minimizes painful procedures that are associated with this outpatient visit. Most children also received oral preoperative sedation with midazolam. Our facility

Fig. 1.

Fig. 2.

Table II. Labial adhesions: recurrence after examination with general anesthesia

Age of referral	*Age at recurrence*	*Symptoms*
20 mo	25 mo	Dysuria
20 mo	37 mo	Twin, required repeat examination with general anesthesia twice
20 mo	34	Dysuria HX of extreme with estrogen
3 y	3 y 4 mo	Responded to estrogen tissue bridge over urethra
3 y 11 mo	5 y 6 mo	Discomfort, responded to estrogen
16 mo	22 mo	Responded to estrogen
15 mo	3 y	Urinary frequency; tissue bridge over urethra; responded to estrogen, topical anesthetic
2 y 9 mo	5 y	Minimal posterior agglutination, no treatment
4 y 6 mo	5 y	No symptoms, posterior agglutination only, no treatment
3 y 6 mo	3 y 11 mo	Abnormal urine stream, required repeat examination under anesthesia
3 y 6 mo	4 y 8 mo	No symptoms, responded to estrogen and topical anesthetic

HX, History.

prohibits medications for "conscious sedation" without monitoring in the outpatient surgical suite.

The remaining patient, after initial office referral, elected to proceed with continued topical therapy after instruction on application techniques (including general traction against the midline by patient or caretaker). This resulted in approximately 50% opening of the adhesions in the vestibule and thinning of the midline so that a translucent area could be noted. Topical anesthesia with EMLA also assisted separation of the adhesions in the office.

It was noted that the patients whose condition required manual separation had adhesions that were thick (generally 3-4 mm in width). Only a tiny opening in the adhesions could be detected. They were separated manually revealing a raw surface with pinpoint bleeding sites. Separation of the adhesions was performed from the base of the clitoral hood toward the posterior fourchette. No attempt was made to relieve any agglutination of the clitoral hood because this is a normal anatomic finding in prepubertal girls and resolves spontaneously after puberty.

After the separation of the adhesions, close inspection of the vestibule and lower one third of the vagina and introitus was performed. After the inspection, a mixture of estrogen cream and Xylocaine Jelly 2% was applied to the adhesion site.

Topical estrogen cream was used nightly for 2 weeks after the procedure. Topical anesthetic jelly was used until patient comfort was obtained; the patients were advised to use sitz baths without soap or detergents. In addition, a bland emollient (eg, white petroleum jelly) was recommended with each toileting or diaper change until the postoperative visit 2 weeks later and nightly thereafter for 4 to 6 months and whenever vulvar irritation occurred.

Office follow-up visits were scheduled 2 weeks after the procedure and thereafter on an as needed basis. Patients and caretakers were told to inspect the area periodically for recurrences. Recurrences were to be promptly reevaluated.

One patient complained of postoperative vulvar discomfort, and an erythematous area developed that was consistent with a fungal infection. This responded to antifungal therapy.

Side effects of therapy. Four of 23 patients reported unpleasant side effects from the estrogen cream. The predominant cream prescribed was a conjugated estrogen cream (although 1 patient had been treated with estradiol cream). All the adverse side effects were reported in patients who used a conjugated estrogen cream. Side effects included erythema in several patients and breast "budding" in 2 patients. Both patients with breast changes noted regression after discontinuation of the estrogen. One of the 2 patients had also reported increased body hair while using the estrogen cream and noted partial regression on discontinuation. No patients reported vaginal bleeding.

Recurrence. Recurrence of labial adhesions after examination with general anesthesia was experienced by nine patients (Table II). Two patients had a second recurrence, and one patient had a third recurrence. All patients were treated initially with topical estrogen cream. Each child who had a recurrence was examined promptly by the investigator, within a few weeks of the noticed occurrence. Two patients required no therapy because of minimal agglutination that occurred in the posterior vestibule that was accompanied by an absence of symptoms. Four of nine patients responded to topical estrogen therapy. Examinations in the office revealed these adhesions involved less of the vestibular opening than previously, appeared thinner, or exhibited translucency in the midline.

Patients with an adverse response to previous topical estrogen were treated with a different estrogen preparation (such as estradiol cream vs conjugated estrogens) and were placed on estrogen therapy for only a 2- to 3-week trial period.

Three of nine patients did not respond to medical therapy of the recurrent adhesions and had recurrent thick adhesions that involved the entire vestibule and eventually required repeat manual separation. One of these three patients required a third manual separation, again with recurrent thick adhesions. This patient was one of the patients who had erythema and vulvar discomfort to multiple estrogen preparations.

Patients who had recurrences were only considered for a repeat examination with general anesthesia because symptoms or urinary symptoms recurred or adhesions were thick and involved >90% of the vestibule.

Two of the four patients with recurrences were twins, both of whom required repeat manual separation after discomfort and erythema with multiple estrogen preparations and the redevelopment of thick adhesions.

It was interesting to note that two of the recurrences appeared as a bridge of tissue that was located across from the urethral opening. Both responded to topical estrogen therapy.

Comment

Patients in this study generally match those classically reported in the literature. All of the patients were <6 years old at examination. Most of the patients were toddlers or in diapers.

No causes or precipitating factors for labial agglutination were identified. Two families had siblings with the same diagnosis; in one family, the siblings were a set twins. There are no separate reports of familial clustering of this process. Similar skin sensitivity, hygienic practices, and possibly other predisposing factors make the possibility likely.

All patients were treated in the traditional fashion with medical therapy that consisted of estrogen cream. Practitioners have been most accustomed to using conjugated estrogen cream, but other preparations (such as estradiol or compounded preparations of estradiol in petrolatum) are less irritating because creams, by definition, contain some alcohol in the base. The alcohol may be the irritant, not the estrogen. Many patients had received a prolonged trial of estrogen, although several weeks is recommended. Prolonged estrogen use certainly contributes to the development of side effects such as breast budding. Failure to respond to estrogen therapy should evoke a discussion of surgical or manual separation.

Treatment of adhesions in children with symptoms remains appropriate, but treatment continues to be controversial in children who are completely asymptomatic, especially younger children in whom there is a longer duration until endogenous estrogen production and therefore perhaps a greater risk of recurrence. Increased educational efforts for clinicians and families combined with periodic evaluation at well child care visits could reduce the number of procedures that are performed only for parental or caretaker concerns.

The patients in this study generally matched those classically reported in the literature. No specific causes or precipitating factors for labial agglutination were identified.

Gynecologists may often receive referrals for the treatment of labial agglutination after failed medical therapy. These patients are noted to have thick adhesions with no thin translucent raphe. These thicker adhesions are more likely to fail to respond to medical treatment and to require manual separation.

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Discussion

Dr C. Kathy Chance, Augusta, Ga. Labial agglutination or adhesions are the postnatal fusion of the labia minora in the midline of varying degrees. It is postulated to be the result of low estrogen levels in the prepubertal child and possibly of a chronic inflammatory process that causes the denudation of the thin surface epithelium of the labia minora that then adhere to one another in the midline because of natural approximation as healing occurs. Its incidence has been estimated at 0.6% to 3.0% but is probably higher because those children with asymptomatic or lesser degrees of adhesion are never reported.[1] The diagnosis is made by the visual inspection of a thin, avascular line in the midline and should not be confused with other vulvovaginopathies (such as ambiguous genitalia, vaginal septi, or imperforate hymen).[2]

Dr Bacon's study population includes 23 children who were referred to her with labial agglutination; none of these children had resolution with topical estrogen treatment provided by the primary care provider. Findings at examination included >90% of the vestibule closed with thick adhesions (3-4 mm), which probably explains the reason that the estrogen treatment failed. Dr Bacon then treated these patients in an outpatient surgical setting with examination with general anesthesia and manual separation of the labia using a cotton-tipped swab and beginning at the base of the clitoral hood toward the posterior fourchette. This approach was successful in all of the study patients, even though a few of the patients had to be separated similarly again at a later date after recurrence and continued failure to respond to topical estrogen.

Dr Bacon agrees with earlier investigators that asymptomatic patients need not be treated and that their parents should be given reassurance because most of these adhesions will resolve spontaneously at puberty.[3] However, the presence of symptoms (including pain, pulling, or complications of the urinary tract) is reason for intervention. Again, topical estrogen application is considered first-line therapy. Conjugated estrogen cream is most commonly used; however, Dr Bacon suggests that estradiol or dienestrol preparations may be better tolerated by the patient.

If adhesions persist, intraoffice manual separation may be appropriate for some patients, with the use of topical anesthetic preparations. If this fails or is not deemed appropriate, Dr Bacon has shown efficacy and safety using an intraoperative setting and a technique for separation of the labia that would certainly decrease pain and emotional trauma in these patients. She also advises prolonged postoperative care that includes the application of topical estrogen for 1 to 2 weeks, followed by application of an emollient for 6 to 12 months, in an attempt to prevent recurrence.

You mentioned the use of oral preoperative sedation with midazolam used with mask anesthesia (without a need for intravenous medication or blood work) as agreed on by your anesthesia department in this study. One of your references is a paper by Jamieson and Ashbury[4] in which they reported success using orally administered midazolam as a short-acting sedative/hypnotic at a dose of 0.75 mg/kg (maximum dose, 20 mg) which required minimal monitoring and resulted in minimal discomfort when combined with EMLA cream and in little recall of the procedure, allowing those children to undergo re-examination at a future date without fear. Do you have any experience with this? In your opinion, is this a reasonable alternative to treatment in an operating room setting, with cost-effectiveness in mind?

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Dr Bacon (Closing). Oral sedation is an excellent alternative to mask anesthesia for manual separation of labial adhesions. Combined with topical anesthesia, it poses little risk to the patient.

Physicians who want to use this protocol are referred to the cited references earlier. The physician also could become familiar with the institution's requirements for the administration of conscious sedation in the outpatient setting. We do not have the required monitoring equipment in our office.